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Systematic review of family functioning in families of children and adolescents with chronic pain

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Abstract

Disturbances in family functioning have been identified in youth with chronic pain and are associated with worse child physical and psychological functioning. Assessment measures of family functioning used in research and clinical settings vary. This systematic review summarizes studies investigating relationships among family functioning, pain and pain-related disability in youth with chronic pain. Sixteen articles were reviewed. All studies were cross-sectional, seven utilized between-group comparisons (chronic pain versus healthy/control) and twelve examined within-group associations among family functioning, pain and/or pain-related disability. Studies represented youth with various pain conditions (e.g., headache, abdominal pain, fibromyalgia) aged 6 – 20 years. Findings revealed group differences in family functioning between children with chronic pain and healthy controls in five of seven studies. Significant associations emerged among family variables and pain-related disability in six of nine studies with worse family functioning associated with greater child disability; relationships between family functioning and children's pain were less consistent. Different patterns of results emerged depending on family functioning measure used. Overall, findings showed that families of children with chronic pain generally have poorer family functioning than healthy populations, and that pain-related disability is more consistently related to family functioning than pain intensity.

Keywords

Child; adolescent; chronic pain; disability; family functioning

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Introduction

Family factors are significantly associated with the physical and psychosocial functioning of children and adolescents with chronic pain.²⁵ These family factors include family functioning and parenting and dyadic parent-child variables that are embedded within the context of the family. Family functioning refers to the social and structural properties of the global family environment. It includes interactions and relationships within the family, particularly levels of conflict and cohesion, adaptability, organization, and quality of communication. Healthy family functioning occurs within a family environment with clear communication, well-defined roles, cohesion, and good affect regulation. In contrast, poor family functioning occurs within families with high levels of conflict, disorganization, and poor affective and behavioral control.¹

Theoretical models, particularly operant behavioral theories and the McMaster model of family functioning have been utilized to describe interactions between adolescents with chronic pain and their parents. Recently Palermo and Chambers introduced a more integrative model to conceptualize family factors in pediatric pain populations.²⁵ The model describes how parenting variables occur within the context of dyadic relationships, and both are embedded in the global functioning of the family environment. Within this model, pain and associated disability have reciprocal influences with factors at each of these levels, and can broadly impact the functioning of the family system. For example, parent-adolescent conflict may reinforce pain behavior, leading to both increased disability and greater stress in the family environment. The model also proposes that individual factors (e.g., coping, psychological functioning) mediate/moderate relationships among family factors and pain and disability. The current review focuses specifically on these pain and disability variables to test the proposed pathways.

Family functioning is an important focus of research in pediatric chronic pain because studies have shown these families report poorer family functioning compared to families with healthy children.³ In addition researchers have found significant associations between family functioning and pain-related experiences. For example, in adolescents with chronic pain, greater family conflict and higher levels of enmeshment have been associated with increased pain-related disability.^{16, 19} Frequency of family conflict in chronic pain populations has also been associated with increased pain occurrence.¹⁵

Although many aspects of family functioning (e.g. conflict, cohesion, organization/structure) have been identified as important in understanding children's experience of chronic pain, there has been limited attention to synthesizing this literature base. Recently researchers conducted a review of measures assessing socioemotional functioning in parents of children with chronic pain.¹² While the review provided important information regarding reliable and valid assessment of parent-level factors, it did not synthesize or report child outcomes in relationship to family functioning. A literature review identifying specific aspects of family functioning that are associated with pain and pain-related disability in children and adolescents is critically needed. Currently it is not clear how specific measures differentially assess domains of family functioning in this population or how specific assessment measures may be associated with different findings. Such a synthesis will also assist in identifying domains of family functioning to serve as targets for future clinical interventions with children with chronic pain and their parents.

The current review targets studies examining measures of general family functioning rather than measures specific to a chronic health condition or disease management. The aims of this review are threefold: 1) to review between-group differences in family functioning between children with chronic pain conditions and healthy youth or normative populations,

2) to examine within-group associations for family functioning with pain, and disability described in Palermo and Chambers' model, and 3) to synthesize the literature examining family functioning in children with chronic pain and present recommendations for future research, clinical interventions, and family functioning assessment.

Materials and Methods

Data Sources

Electronic searches of the Cochrane Database of Systematic Reviews, Medline, EMBASE, CINAHL, and PsychINFO were conducted by a Library Information Specialist familiar with the field. The time period for the searches was from each database's inception through December 2008. Subject headings and MeSH terms included "pain," "parent," "family," as well as specific terms for pain conditions and type of parent and family variables such as "child," "adolescent," "chronic pain," "disability," "family function," "family relation," "parent-child relations," "environment," "abdominal pain," "headache," "migraine," "sickle cell disease" and "arthritis." Nine additional articles were obtained from hand searching of reference sections. No attempt was made to locate unpublished material or contact researchers for unpublished studies (e.g., dissertations or conference proceeding abstracts).

Study Selection

To be eligible, articles had to meet the following criteria:

- Published in a peer-reviewed journal through December 2008.
- Focused primarily on children/adolescents.
- Included youth who were experiencing chronic pain (as defined by individual study criteria).
- Evaluated the relationship among family functioning and children's pain characteristics and/or pain-related disability.
- Used an established self-report measure of family or parent factors (e.g., documented reliability/validity, standardized questionnaires). Studies using single item questions, systematic reviews, practice guidelines, commentaries, or articles focusing on the initial validation of a family functioning measure were excluded from the review.
- Published in English
- Was a descriptive study that did not include an intervention intended to modify family functioning and/or pain/disability.

Review Process

Each database was searched by an independent reviewer (SH) to perform an initial screening for relevancy using study titles and abstracts. A random sample of abstracts was reviewed by another reviewer (JS) with 96% agreement on inclusion in the study. For each database, the reviewer (SH) recorded details of number of studies found, number meeting inclusion criteria, and number excluded.

Data Extraction

Two reviewers (TP, AL) extracted data from all relevant articles using a standardized form. The form summarized basic information about the study including author, year, and description of the target population including age and relevant clinical diagnoses. The form

also outlined outcomes measured and results of the study. Following data extraction, each eligible article was again reviewed in full by two authors (TP, JS).

Results

Of 131 identified articles, 16 met the inclusion criteria and were included in the review. These 16 studies were conducted between 1993 and 2008; the majority (13/16) published after 2000. Most were conducted in North America (12) with others being conducted in European countries. All studies included in this review used cross-sectional study designs. See Table 1.

Family Functioning Measures Included

Studies included in this review used measures of general family functioning and instruments designed to assess specific components of family functioning (e.g., conflict, cohesion, communication, enmeshment). “Family” was defined as the family unit and corresponding subsystems, a definition that had been used in a previous review.¹ Descriptions of the measures and their psychometric properties are listed below. In addition, the evidence base available for each measure based on quality ratings conducted by Alderfer and colleagues¹ is included where available.

The McMaster Family Assessment Device (FAD)²¹ is a 60-item self-report measure that assesses seven dimensions of family functioning: problem solving, communication, roles, affective responsiveness, affective involvement, behavioral control, and general functioning.⁸ Items are rated on a 4-point Likert scale, with lower scores indicating better functioning. Sample items include: “Making decisions is a problem for our family,” “We don’t get along well together,” and “We confide in each other.” Studies in the current review utilized the FAD in its entirety or exclusively the General Functioning subscale. Established cutoff scores are available to categorize families as demonstrating healthy versus unhealthy family functioning. The reliability and validity of this measure has been well documented.²² The FAD received a rating of “well-established” in Alderfer and colleagues’¹ recent review of evidence-based assessment of family measures.

The Family Environment Scale (FES)²³ is a 90-item measure designed to assess the social and environmental characteristics of family functioning, particularly interpersonal relationships, personal growth and family structure/organization. The measure is based upon a three-dimensional conceptualization and has ten subscales which include: the Relationship dimension (Cohesion, Expressiveness, Conflict), the Personal Growth dimension (Independence, Achievement, Orientation, Intellectual-Cultural Orientation, Active-Recreational Orientation, Moral-Religious Emphasis), and the System Maintenance dimension (Organization, Control). Sample items include: “Family members often keep their feelings to themselves,” “We feel it is important to be the best at whatever you do,” “Family members have strict ideas about what is right and wrong.” If the scale is used in its entirety, families can then be grouped along the three conceptual dimensions (Relationship, Personal Growth, and System Maintenance). There are three available versions of the FES, the original scale, the Family Relationship Index, and the Kronenberger and Thompson factor-analytic component scoring system. The reported internal consistency of subscales is low to moderate (.61 – .78) and test-retest reliabilities range from low to high (.52 – .91).²³ Two versions of the FES (Original scale; Kronenberger and Thompson) received a rating of “approaching well-established” and the Family Relationship Index was rated “well-established” in Alderfer and colleagues’¹ evidence-based review.

The Family Adaptability and Cohesion Scale (FACES III)²⁴ is a 20-item measure designed to assess two dimensions, adaptability and cohesion, by targeting seven areas of family

function: problem solving, behavior control, roles, affective involvement, affective responsiveness, and communication.³⁰ Sample items include: “Our family does things together,” “Family members avoid each other at home,” and “It is easier to discuss problems with people outside the family than with other family members.” Internal consistency has been demonstrated as fair.²⁴ Alderfer et al.’s¹ review of evidence-based assessment of family measures rated the FACES as “approaching well-established”.

The Stress Index for Parents of Adolescents (SIPA)³⁶ is a 112-item measure originally developed to assess stress in parents of healthy adolescents. The first 90 items are scored on a 5 point rating scale (1 Strongly Disagree – 5 Strongly Agree), and the final 22 items assess exposure to stressful life events with Yes/No response options. Sample items include: “My child talks to me about problems,” “My child thinks I do not love him/her,” and “It is easy for me to understand what my child wants or needs.” The four domains assessed are general symptomatology of both caregiver and child, the adolescent-parent relationship, and the caregiver’s stressful life events. The single study that utilized the SIPA in the current review used the scores on the adolescent-parent relationship domain (APRD) exclusively. The SIPA has demonstrated excellent internal consistency and test-retest reliability in validation studies with normative samples of parents and adolescents.³⁶ The SIPA was not evaluated in Alderfer et al.’s¹ review of evidence-based assessment of family measures.

The Bath Adolescent Pain Questionnaire (BAPQ)⁷ is a multidimensional questionnaire with both parent and adolescent versions that includes 61 items to assess how adolescents’ functioning is affected by chronic pain. The entire measure has seven subscales (social functioning, physical functioning, depression, general anxiety, pain specific anxiety, family functioning, and self-perception of development) with item responses on a 5 point rating scale ranging from “never” to “always”. Sample items include: “Family life is stressful,” “There are fights between members of my family,” and “I feel close to other family members.” Results from analyses that included responses on the 12 item family functioning subscale were included in the current review. The BAPQ has established reliability and validity in samples of adolescents with chronic pain.⁷ The measure was not evaluated in Alderfer and colleagues’¹ review of evidence-based assessment of family measures.

Assessment of Pain and Disability in Included Studies

Reviewed studies assessed pain and pain-related disability using a variety of assessment measures. Assessment tools varied in terms of length (e.g. single items versus multi-item measures) and available evidence-base. Pain assessment included measures of intensity, pain frequency, and number of pain days on likert or numerical rating scales. The FDI³⁸, CALI²⁷, amount of health care utilization, fatigue, and disease specific measures (e.g., Modified Fibromyalgia Impact Scale) were used to assess disability. For the purposes of this review the definition of disability was broad so that the maximum number of studies could be included.

Characteristics of Studies Included in Review

Included studies represented a range of pain conditions including recurrent headaches/migraines (n = 2), abdominal pain (n = 1), sickle cell disease (n = 1), fibromyalgia (n = 2), arthritis and musculoskeletal pain (n = 2), and mixed conditions (n = 8). Seven studies employed a healthy group and/or another chronic illness group or a normative population for comparisons. Only one study used community-based recruitment while the remaining studies utilized clinical samples. The majority (n=15) of studies focused on youth who were school-age (6 years) to late adolescence (18 years). A single study, included participants up to 20 years of age.³⁴ Sample sizes of the studies ranged from 45–183 with 88 being the median number of participants.

The majority of studies included in this review used either the FAD (n = 6; total measure or General Functioning subscale only) or the FES (n = 6) to examine between group differences and associations among family factors and pain and functional limitations. The three other measures used in studies included in this review were used less frequently. The Bath Adolescent Pain Questionnaire (BAPQ) was used in two studies by the same investigative team.⁷ Both the SIPA (adolescent-parent relationship subscale only) and the FACES were used in single studies. See Table 1.

Outcomes of Studies

Group Differences—The primary aim of seven studies was to evaluate group differences in family functioning of children and adolescents with chronic pain compared to healthy controls. Depending on the measure used, family factors were assessed by reports of general family functioning, cohesion, conflict, organization, relationship distress, expressiveness and independence. Collectively, results of these studies revealed group differences in five of the seven studies, with families of youth with pain conditions demonstrating significant differences between other pain groups, healthy youth, or when compared to normative data. Specific findings are described below and additional details are presented in Table 1.

In the two studies using the FAD to assess group differences, differences in family functioning were found between youth with chronic pain and control groups. On the General Functioning subscale, Antilla and colleagues³ found that children with headache had worse family functioning and more family problems compared to healthy controls. In participants with either abdominal pain or headache, Liakopoulou-Karis and colleagues¹⁸ reported that families of children with abdominal pain showed significantly greater family disturbances than controls across all domains. In addition, participants with headache had worse family functioning compared to controls exclusively on the domains of Behavior Control and General Functioning. There were no differences, however, between families of youth with headache and abdominal pain.

The findings from the four studies that used the FES to assess group differences showed greater variation. Comparing adolescents with organic and nonorganic abdominal pain and healthy controls, Kaufman and colleagues¹⁴ did not find any group differences on family environment variables. The other three studies revealed significant group differences although one was in the unexpected direction. Supportive of their hypotheses, Conte and colleagues⁵ found that when comparing children with fibromyalgia, arthritis, and healthy controls, participants with fibromyalgia had significantly less family cohesion, less organization and more conflict compared to the other groups. When rating themselves, parents of children with fibromyalgia also reported higher levels of anxiety and depression, and overall poorer psychological adjustment. Similarly Kashikar-Zuck and colleagues¹³ found that adolescents with fibromyalgia had poorer overall family environments compared to controls; particularly higher levels of conflict, less cohesion, and less organizational structure.

In contrast to these findings, in Schanberg and colleagues'³⁴ study of adolescents with fibromyalgia and their parents, both parents and youth with pain reported significantly less conflict than normative samples of parents of healthy and distressed adolescents. When compared to a normative sample of distressed families, parents and youth with fibromyalgia endorsed greater cohesion, intellectual-cultural orientation and moral-religious emphasis. Compared to their parents, adolescents with fibromyalgia perceived the family environment as more cohesive.

Reid and colleagues'²⁹ study comparing group differences in family functioning in children with fibromyalgia, juvenile rheumatoid arthritis, and healthy participants was the single

study to use the FACES. Results did not show any differences in family functioning domains of adaptability or cohesion amongst the three groups.

In sum, four of seven studies revealed youth with chronic pain have greater disturbances in family functioning across multiple domains (e.g., less cohesion and organization, more conflict) when compared to healthy control groups or population norms^{3, 5, 13, 18}. Of the three remaining studies, two did not reveal any group differences^{14, 29} and a single study found that adolescents with pain and their parents endorsed better functioning than normative samples.³⁴

Associations among Family Functioning and Children's Pain and Disability—

A second aim of this review was to examine the within-group associations among family functioning and ratings of children's pain and pain-related disability. Several studies showed significant associations among pain, disability, and family functioning, with the relationship between increased disability and increased family dysfunction being the most consistent. Associations between pain and family functioning were equivocal, and in some studies in the unexpected direction. Specific findings are described below.

The FAD was used to assess relationships among pain, functional limitations and family functioning in three studies, with two showing significant associations. Palermo and colleagues²⁶ study of adolescents with recurrent headache revealed that adolescents with healthy family functioning had lower levels of depression, less functional impairment, and lower pain frequency and intensity compared to adolescents with unhealthy family functioning. Iobst and colleagues¹¹ found an association between family functioning and fatigue (as assessed by days feeling tired) in patients with juvenile rheumatic disease, with higher levels of fatigue associated with worse family functioning. Findings did not reveal significant associations between FAD scores and number of pain days.

Mitchell and colleagues²² found no relationship among FAD scores and disease severity or health care utilization in a study of children with sickle cell disease. However findings revealed several aspects of family functioning were associated with children's coping strategies and levels of negative thinking (as rated by their parents). Specifically the use of active coping strategies was related to higher scores on Problem Solving, Communication, and General Functioning domains. Lower levels of negative thinking were related to higher scores on Communication, Affective Involvement, Roles and Behavioral Control domains.

Five studies utilized the FES to assess relationships among family environment and both pain and functional limitations. In children with migraine headache or abdominal pain, Logan and Scharff²⁰ reported that when controlling for pain intensity, both conflict and family enmeshment predicted functional disability. In terms of group differences, family environment moderated the association between pain and disability in youth with migraine but not in youth with abdominal pain.

In Schanberg and colleagues³⁴ study of adolescents with fibromyalgia, adolescent report of greater intra-family control was correlated with higher pain scores. Youth who reported higher levels of family expressiveness also endorsed less pain and had lower scores on a scale of fibromyalgia-related disability. On parent report, more positive reports of family functioning (active-recreational and intellectual-cultural orientation domains) were correlated with less child psychological distress and better physician-reports of global health. Of note, analyses comparing parent and child agreement on family functioning measures revealed that more incongruence between parent and child report of family environment was associated with greater impairment. Kashikar-Zuck and colleagues¹³ also studied associations among family functioning and pain, and pain-related disability in a

sample of adolescents with fibromyalgia using the FES. Findings indicated that controlling for pain, the total model (family environment, adolescent and marital variables) accounted for 48.1% of variance in functioning. Family environment variables alone did not predict functioning.

Contrary to hypotheses, Ross and colleagues³¹ found that when controlling for disease and psychological variables, greater harmony in the family environment was associated with higher reports of pain in children with juvenile rheumatoid arthritis. This finding is similar to that reported by Logan and colleagues¹⁹ who also noted that pain and family distress (as assessed by the SIPA) were inversely related. Logan et al.'s¹⁹ study also revealed that the relationship between pain severity and functional disability was moderated by parent-adolescent relationship distress; specifically pain severity was more closely related to functional disability when relationship distress was lower.

Scharff and colleagues³⁵ used the FES to assign children with chronic pain to one of three clusters: Distressed/Low Functioning (DLF), Interpersonally Distressed (ID), or Family Dysfunction (FD) based upon coping style, level of distress, and social/behavioral functioning. Neither pain level nor duration was associated with group assignment; however, significant differences emerged on the Cohesion and Accepting Responsibility subscales, with participants in the FD cluster reporting less cohesion and greater responsibility taking than the other two groups. The DLF cluster reported significantly more functional disability than both other groups, and the FD group was significantly higher on functional disability than the ID group.

Finally, both Gauntlett-Gilbert et al.⁹ and Eccleston et al.⁷ used the BAPQ to assess relationships among pain, disability and family functioning within mixed chronic pain samples. Gauntlett-Gilbert and colleagues'⁹ study revealed poorer family functioning was significantly correlated with greater functional disability, lower school attendance, more depression, general anxiety, and pain specific anxiety. When family functioning was included in a multivariate model that included depression, anxiety, and pain specific anxiety, the total model accounted for 41% of the variance in functional disability, however family functioning alone was not a significant predictor. Eccleston and colleagues'⁷ study revealed significant correlations among poorer family functioning and higher pain specific anxiety, and worse social functioning. Controlling for pain intensity, family functioning was a significant predictor of children's emotional adjustment.

In sum, six studies revealed hypothesized associations between family functioning and pain-related disability (as either a direct effect or part of a regression model) with better family functioning associated with less disability, or higher disability scores associated with more family dysfunction.^{7, 9, 11, 20, 26, 13} Two studies showed no associations between functional disability and family factors^{19, 30} and in one study, no association was found between family functioning and health care utilization variables.²²

Studies examining associations between family functioning and pain were more variable both in significance of findings and in the direction of effects. One study found a relationship between pain and family factors³⁴ and two studies showed relationships between family factors and pain specific anxiety.^{7, 9} Two studies had unexpected associations with pain and family functioning, with better family functioning associated with higher pain or more family distress correlated with lower pain.^{19, 31} Finally, a single study showed paradoxical associations between pain and family factors, with greater intra-family control associated with higher pain but greater family expressiveness associated with lower pain ratings.

Discussion

This review synthesized findings from 16 studies describing functioning in families of children with chronic pain. Four of seven studies revealed poorer functioning in families of youth with chronic pain compared to healthy adolescents or population norms. These findings extend previous research indicating that families of children with medical conditions have higher stress and lower socioemotional functioning than families with healthy children.^{6, 10, 37} The majority of studies examining pain-related disability revealed poorer family functioning was associated with greater disability. Six of nine studies revealed hypothesized associations between family functioning and pain-related disability (as either a direct effect or part of a regression model) with better family functioning associated with less disability, or higher disability scores associated with more family dysfunction.^{7, 9, 11, 20, 26, 13} This supports Palermo and Chambers' ²⁵ model describing interactions among family-level variables and pain-related disability.

While many studies showed hypothesized between-group differences in family functioning as well as within-group associations between family functioning and disability, equivocal findings also emerged. The most striking was variability in associations between pain and family functioning, with studies showing better family functioning associated with more pain, less pain, or both^{19, 31, 34} While unexpected, findings showing family harmony and cohesion were associated with higher pain suggest that some pain experiences may unite parents and adolescents and reduce relationship distress. It is possible that an environment high in family harmony leads parents to be more responsive to children's pain. Similar protective effects were shown by Palermo and colleagues with higher family functioning associated with less depression and functional impairment.²⁶ While these protective effects were identified, the majority of research to date has focused on negative outcomes associated with poor family functioning. Future research should explore family functioning as a possible protective factor.

Variability in findings was likely associated with the outcome measures used. The majority of studies used measures with established reliability and validity (e.g., FDI) however assessments of pain and pain-related disability were not consistent across studies. It will be important for future research to assess multiple characteristics of pain and functioning. For example, the majority of studies examined pain intensity; only two studies looked at family factors in relation to pain frequency or duration. Chronic pain is taxing on families and associations with pain duration may be particularly salient.

Additional measurement and design issues should be considered when interpreting these results. First, the family functioning measures included varied in scope, domains assessed, and quality (per Alderfer et al.'s rating¹). The FACES-III, FES and the FAD were developed to assess broad dimensions of family functioning. Advantages of these measures are they are broad, cover multiple domains, and allow for group comparisons. However, most general measures have not been validated for use with pain populations and lack normative data, limiting conclusions that can be drawn. The BAPQ was the only measure developed for families of children with chronic pain. Only two studies^{20, 35} reported reliability (e.g., Cronbach's α) of the family functioning measure in their pain sample, highlighting gaps in psychometric data on family functioning measures.

Evidence-base for the included measures may have also impacted results. A single study utilized the FACES³⁰, a measure with limited use recently because it was developed based on a dimensional model that lacks empirical support.²⁸ The measure was previously rated "approaching well-established"¹ but lack of associations in the current review may reflect measure limitations.

Of the six measures reviewed, the FAD emerged as the most appropriate tool for assessment of family functioning in pediatric pain populations. Compared to the FES, the FAD had more consistent results with four of five studies showing significant between-group differences or associations between family functioning and disability. Studies using the FES had findings in both expected and unexpected directions, within and across studies. In addition, the FAD is shorter and includes a “General Functioning” subscale that can be scored independently. This recommendation is supported by the large evidence-base for use of the FAD with pediatric samples (e.g., cancer, asthma, diabetes) and strong psychometrics (rating of “well-established”).¹ As the only measure designed for use for adolescents with chronic pain, the BAPQ shows promise. Both studies utilizing the BAPQ revealed associations among family functioning, pain and pain-related disability,^{7, 9} and future studies will provide greater evidence-base.

While this review provides an important synthesis of family functioning in chronic pain populations, limitations should be noted. First, the inclusion of only 16 studies limits generalizability; although reflective of the current state of the literature additional research is clearly needed. Cross-sectional design, small samples, and lack of comparison groups are important limitations in this field. None of the studies used longitudinal designs so temporal relationships between variables could not be assessed. Future studies may clarify changes in parent-child and family relationships in the context of pain over time. Prior research has shown that pain prevalence increases during adolescence,^{17, 32} and it will be important to determine whether family dynamics or parent-child interactions are associated with these changes. In addition, large case-controlled designs are needed to clarify differences between healthy and pain populations.

This small group of studies was also variable in regards to children’s pain conditions and family demographics (e.g. composition, income, ethnicity) and these factors may have impacted the findings. Larger sample sizes are needed to clarify how additional family factors are relevant to particular pain conditions and demographic groups. A final limitation is that only six studies included reports of family functioning by both parents and children. Recent family functioning literature has emphasized the importance of obtaining information from multiple sources.⁴ The importance of examining congruence is demonstrated by Schanberg et al.’s³⁴ study which revealed that degree of incongruence on reports of family functioning, not ratings themselves, predicted impairment. Obtaining both parent and child reports is important before making conclusions regarding the associations among family factors and pain and related disability.

Recommendations for Future Measurement Research

While a variety of family functioning measures are available, additional research on the psychometric properties and clinical utility of established and new measures is necessary. Additional reliability and validity information is necessary to establish norms and clinical cutoffs in pain samples. Measures need to be evaluated for predictive validity and responsiveness to change. Studies examining how family functioning changes over time as adolescents mature are lacking; and data are needed concerning how family functioning changes in concert with children’s pain and functioning.

Clinical Implications and Interventions

A variety of measures are available for assessment of functioning in families of children with chronic pain, and clinical assessment of family factors can be used to identify families most at risk for poor family functioning. Identifying these families can facilitate referrals for

much needed family interventions, assist with treatment planning, and potentially lead to improved outcomes.

To date, interventions for families of children with chronic pain have targeted individual rather than family variables, and have focused on operant strategies (e.g., reinforcing adaptive coping, discouraging maladaptive pain behaviors).^{21, 33} These individual parent and child behaviors often fail to take into account the role that dyadic relationship and family variables (e.g. conflict, communication) may play in pain and pain-related disability. Interventions targeted at parent-child communication strategies to reduce pain and disability is an important future direction.

Perspective

This review highlights the importance of family factors in pain-related disability in youth with chronic pain. Results suggest that family-level variables may be an important target for intervention. Family functioning measures showed significant variation and researchers should take this into account when selecting instruments for use in research and clinical settings.

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Table 1

Characteristics of included studies (n=16)

Author, year, country	Sample Participants (group, sample size)	Participant Age Range	Family Functioning Measure; Reporter	Study Outcomes	
				Group Differences	Pain and Pain-Related Disability
Anttila et al. (2004), Finland	Community based sample Migraine (n=59), Tension HA (n=65), Control group (n=59)	Mean =12.6 yrs (sixth grade)	FAD - General Functioning subscale; Parent report only	Youth with both tension headache and migraine had worse family functioning than healthy controls (p=. 02).	
Conte et al., (2003), USA	Clinic based sample PJFS (n=16), JIA (n=16), Control group (n=16)	10-17 years	FES; Parent and child report	Compared to arthritis and no pain groups, both children with PJFS (p=. 03) and their parents (p=.01) reported less family cohesion. Children with PJFS reported more organization (p=. 04), and their parents more conflict (p=.05) and intellectual/cultural orientation (p=.01) than both other groups.	
Eccleston et al., (2008), UK	Clinic-based sample Mixed chronic pain sample (n=100)	11-18 years	BAPQ - family functioning subscale; Child report only		Family functioning was correlated with pain specific anxiety (r=.36, p<.001), and social functioning (r=. 31, p<.01). Controlling for pain, family functioning was a significant predictor of children's emotional adjustment (β =.26, p<.01).
Gauntlett- Gilbert & Eccleston, (2007), UK	Clinic-based sample Mixed chronic pain sample (n=110)	11-18 years	BAPQ - family functioning subscale; Child report only		Family functioning was correlated with functional disability (r=.20, p<. 05), school attendance (r=.29, p<. 05), depression (r=.52, p<.001), and anxiety (r=.43, p<.01). A multivariate model (depression, anxiety and family functioning) accounted for 41% of the variance in functional disability (p<.001).
Iobst et al., (2007), USA	Clinic-based sample Juvenile rheumatic disease (n=82)	6-18 years	FAD - General functioning subscale; Parent report only		Days feeling tired predicted worse family functioning (β =-.55, p<.01) No significant associations between FAD scores and # of pain days.
Kashikar- Zuck et al. (2008), USA	Clinic-based sample PJFS (n=47), Control group (n=46)	12-18 years	FES; Parent and child report	Adolescents with fibromyalgia had significantly lower scores on the Family Relationship Index than healthy controls (p=.0002). Families of adolescents with pain reported a less supportive (p=.018) and more conflicted (p=.002) family environment.	Controlling for pain, the total model (family environment, adolescent and marital variables) accounted for 48.1% of variance in functioning. Family environment variables alone did not predict functioning.

Author, year, country	Sample Participants (group, sample size)	Participant Age Range	Family Functioning Measure; Reporter	Study Outcomes	
				Group Differences	Pain and Pain-Related Disability
Kaufman et al. (1997); USA	Clinic-based sample Organic RAP (n=25), Non-organic RAP (n=24), Healthy control group (n=19)	12–16 years	FES; Parent and child report	Adolescents with RAP scored below FES normative means on expressiveness, independence, intellectual- cultural orientation (p<.01). Maternal report on moral religious emphasis and control scales by RAP group scored significantly above the normative means (p<.005). No significant differences between organic versus non- organic bowel disease.	
Liakopoul ou-Kairis et al., (2002) Greece	Clinic-based sample RAP (n=38), HA (n=31), Control group (n=60)	8–13 years	FAD; Parent report only	Families of children with RAP had worse family functioning compared to controls on all domains (p<.05–.001). Families of children with RAP had lower scores on domains of Behavior Control and General Functioning (p<.001). Differences between headache and RAP groups were not significant.	
Logan, et al. (2006); USA	Clinic-based sample Adolescents with mixed chronic pain and their parents (n=112)	13–18 years	SIPA - Adolescent-parent relationship domain subscale; Parent report only		Greater relationship distress was associated with lower pain intensity scores ($r=-.24$, $p<.01$). No relationship between FDI score and adolescent and parent relationship distress.
Logan & Scharff (2005); USA	Clinic-based sample Migraine HA (n=48), RAP (n=30)	7–17 years	FES; Parent report only	Family environment moderated relationship between pain and disability in migraine (p<.001) but not in RAP group	Controlling for pain intensity, family conflict (p<.01), organization (p<.01), independence (p<.05) and conflict (p<.001) predicted functional disability
Mitchell et al. (2007); USA	Clinic-based sample SCD (n=48), Caregivers (n=53)	7–13 years	FAD; Parent report only		No relationship between FAD scores and other medical, pain or health care utilization variables Family functioning was associated with children's coping strategies and level of negative thinking ($r=-.31$ to $-.37$)
Palermo & Putman (2007); USA	Clinic-based sample Recurrent HA (n=49)	11–16 years	FAD; Parent and child report	Adolescents with healthy family functioning had less depressive symptoms (p=.002), functional impairment (p=.01), a trend toward less pain frequency (p=.08) and pain intensity (p=.09) compared to adolescents with unhealthy family functioning	Lower levels of adolescent autonomy ($r=-.53$, $p<.01$) and worse family functioning ($r=-.32$, $p<.01$) were correlated with greater impairment. Adolescent autonomy ($\beta=-.38$, $p<.05$) and family functioning ($\beta=-.34$, $p<.051$) predicted impairment.

Author, year, country	Sample Participants (group, sample size)	Participant Age Range	Family Functioning Measure; Reporter	Study Outcomes	
				Group Differences	Pain and Pain-Related Disability
Reid et al., (2005), Canada	Clinic-based sample JIA (n=15), Fibromyalgia (n=15), Control group (n=15)	10–17 years	FACES; Parent and child report	No significant group differences in family functioning	Family functioning is not related to functional disability
Ross et al.; (1993); USA	Clinic-based sample JRA (n=56)	10–17 years	FES; Parent report only		Greater family harmony was associated with higher reports of pain ($r=.35$, $p<.01$)
Schanberg et al.; (1998) USA	Clinic-based sample Parents (n=29) of children with JPFS (n=21);	10–20 years	FES; Parent and child report	Parents of children with JPFS reported lower levels of family cohesion ($p<.001$), and both parents and children reported less conflict ($p<.05$) compared to healthy parents and adolescents. Compared to distressed group, both parents ($p<.01$) and children with JPFS ($p=.001$) reported less conflict.	Child-report of family expressiveness was associated with less pain ($r=-.49$, $p<.02$), lower levels of impairment ($r=-.55$, $p<.04$), and greater physical function ($r=-.46$, $p<.04$). Greater incongruence between parent and child report of family environment was associated with greater impairment ($r=-.57$, $p<.003$).
Scharff et al. (2005); UAS	Clinic-based sample Mixed chronic pain group (n=117)	8–17 years	FES – cohesion subscale; Parent report only		Neither pain level nor duration was associated with group assignment Youth in Family Dysfunction cluster reported less cohesion ($p<.001$) and greater responsibility taking ($p<.005$) than the other two groups.

Note: Sickle Cell Disease = SCD; Headache = HA; RAP= Recurrent Abdominal Pain; Idiopathic Musculoskeletal Pain = IMP; Juvenile Idiopathic Arthritis = JIA; Primary Juvenile Fibromyalgia Syndrome = PJFS; Family Assessment Device = FAD; Functional Disability Inventory = FDI; Stress Index for Parents of Adolescents = SIPA; Family Environment Scale = FES; Family Adaptability and Cohesion Scales = FACES; Bath Adolescent Pain Questionnaire = BAPQ; USA = United States; UK = United Kingdom